

The Intersexed Patient

ROBERT J. STOLLER, M.D., and ALEXANDER C. ROSEN, Ph.D., Los Angeles

A MOST DIFFICULT and rare, although most interesting, problem for a physician is that of the "intersexed" patient—that is, one in whom there is demonstrable abnormality, anatomically and/or biochemically, especially in the urogenital system.* When the abnormality is discovered in infancy, there is the problem of how to raise this person; later in life there are the problems of whether or not to assist the patient in a change of sex. It is well known that experts (Cappon¹ and Money,⁵ for example) fundamentally disagree on these issues. The physician's determination as to the most appropriate sex is vital in shaping the patient's future. Fortunately, this decision is approached with great caution.

Kiefer⁴ has defined sex as "the overall state of body and mind by which the individual conforms to the masculine or feminine standards of normality in the named sex-determining factors [chromosomes, gonads, hormones, sex organs, and psychic pattern]. It is an algebraic summation of these factors in which no one factor supersedes the others." This is the sense in which the word *sex* will be used in this communication.

Freud, and analysts who followed, demonstrated the presence of both masculine and feminine qualities in the character structure of all people. Freud explained this as the psychological manifestation of a biological bisexuality,³ which researchers of recent years have fully demonstrated. The anatomists have shown the presence of vestigial female reproductive organs in the male, and vice versa; embryologists have traced back the presence of these vestigial organs to an undifferentiated state of anatomical sex early in the embryo; by tampering with the early formed embryo, experimental biologists have changed various aspects of its somatic sex from that which had been chromosomally determined; endocrinologists have shown admixtures of both "male" and "female" hormones in both sexes. There has been increasing interest in unusual anatomical and endocrinological anomalies usually grouped under the wide heading of pseudohermaphrodites.

From the Department of Psychiatry, University of California School of Medicine, Los Angeles.

Presented before the Section on Psychiatry and Neurology at the 88th Annual Session of the California Medical Association, San Francisco, February 22 to 25, 1959.

*We are not considering those cases of homosexuality, transvestism, and other related perversions where no etiological (or even concomitant) anatomic or biochemical changes have been shown and where early childhood relationships with parents have been clearly found to be etiological.

• There are at present two opposing points of view on problems of dealing with the intersexed patient (not the typical homosexual or transvestite) who has clearcut anatomical or biochemical qualities of the opposite sex. The first is that in the growing child or adult coming for treatment, the sex the patient should adopt is the summation of somatic sex. The other is that the sex role should be assigned according to the predominant psychological identification already present.

A case history of a middle-aged pseudohermaphrodite, castrated in youth but raised from birth as a female and living thus in "homosexual" relations with women until examined and interviewed at UCLA Medical Center is presented to illustrate the psychological problems in sexual identity with which the patient had to cope.

Psychiatric investigation revealed how confused the patient's sex identity was until treatment by a team consisting of psychiatrist, psychologist and endocrinologist permitted the patient, even at so late a date, finally to establish what his gender is. The patient was able, despite early rearing as a female and a castrating operation, to swing to a more masculine identification. This was possible because of some uncertainty of sexual role from an early age.

Persons with such anomalies may have clearly defined external genitalia of one sex, yet have disturbances in endocrine function which would be commensurate with that sex and with gonads of the opposite sex.

To which sex should these patients be assigned? It is no longer possible simply to assign a person to sex according to the appearance of the external genitalia. Although the vast majority of persons clearly fall into one category or another, the unusual case is much less clear. Instead of using a single criterion as the determining factor in such assignment, recent investigators have been concerned with a number of variables. These are usually divided into two categories, the somatic: (1) chromosomal sex, (2) gonadal sex, (3) hormonal sex, (4) external and internal genitalia, (5) secondary sexual characteristics, (6) body habitus; and the psychological, that is, the sexual identification the person has made.

Two opposing points of view, both based on seasoned research and inquiring thoughtfulness, appear in the literature. The first is exemplified by Cappon,¹ who, in a report on a series of 17 intersexed patients, concluded: "When all the components are

added up, if the physical person has one gender, the mental person has the same gender. It was concluded that they must have a common source. . . . It also follows that sex assignment and rearing should always be in the direction of the preponderant somatic sex. . . . We advocate correcting any area in upbringing and in physiology and anatomy always in the direction of preponderant somatic sexuality to the extent of possibility and as soon as possible. . . ."

An opposing viewpoint is expressed by Money and the Hampsons,⁵ who were of the opinion that somatic gender is much less significant than certain psychological measurements in determining a patient's sex and in thus influencing the physician's decision as to which way to help the patient direct his development.

"From the sum total of hermaphroditic evidence," these investigators said, "the conclusion that emerges is that sexual behavior and orientation as male or female does not have an innate instinctive basis. . . . Sexuality is undifferentiated at birth and . . . becomes differentiated as masculine or feminine in the course of various experiences of growing up. . . . Though gender imprinting begins by the first birthday, the critical period is reached by about the age of 18 months. By the age of two and a half years gender role is already well established."³ Their conclusions were based on the study of 105 intersexed patients.

In another communication the same investigators said that "once a person's gender role begins to get well established, an attempt at its reversal is an extreme psychological hazard."⁶ They expressed belief that in the neonate and young infant, sex assignment is best made on the basis of external genitalia, and later hormonal and other treatment can be given as indicated. For older children, they strongly recommended that the child be left in the same sex as that originally assigned.

We have studied in a research-therapeutic relationship a patient with very anomalous sexual identifications. The patient was a 50-year-old, white, single person who presented herself* as a "butch"† when first referred to the Department of Psychiatry for research study. The patient was seen about once a week for two years.

At the time she was first seen, the patient considered herself a woman and had been brought up as a female from birth. Although some peculiarity of her external genitalia was noted by the physician who delivered her, no question was raised about her sex. The parents were told she was a girl, the birth certificate issued was for a female and she was ap-

propriately named. From birth on, she was considered a girl by her parents, neighbors, and friends and was treated as such. In her dress, the way she wore her hair, her mannerisms and companions, she was treated as and felt herself to be a girl. However, as she grew, she developed external genitalia consisting of a "clitoris" longer than the average girl's but shorter than a boy's penis, with first degree hypospadias, swollen "external labia" and a pencil-width opening an inch or so in depth between the labia.

Family history indicated that at least five other members of the patient's maternal family, and her mother and sister, had anomalies in genital structure.

The patient considered herself to have been a very active child and to have preferred boys' games as far back as she could remember, although she was given dolls and dishes. "I was always thinking of adventurous things that took nerve and daring, like driving racing cars and speed boats, to learn about guns and hunting—but I always thought of myself as a woman doing these things, not as a man."

The patient's mother was described as a meticulous and shy woman, pessimistic and cold, without signs of affection. The patient had no memory of ever having been held or comforted by her mother and felt that her mother wanted neither herself nor her sister. Her mother never enlightened her about sexual matters. Twice during the patient's childhood, her mother was hospitalized for mental illness, for about three weeks each time. The patient said she had noticed no abnormality and did not know why her mother went to the hospital.

She felt much closer to her father, considering him a kind, harassed man who was unable to cope with his wife's sexual and emotional frigidity. He passively retired from dealings with his wife and to a certain extent from the upbringing of the children. He was as restrictive of any conversation regarding sex as was her mother.

Relations with her sister are affectionate. She felt that her sister had been even more shy and retiring than she was because of the peculiarities of their sexual development. For a number of years they lived together. Although they worked and mingled with people at work, they tried whenever possible to isolate themselves from others. The sister was tall, muscular, heavy-boned and had a heavy beard, although thinking herself a woman. The sister had the same physical malformations the patient had; she was considered a female by the delivering physician and was so named and reared.

Sexual History

Despite the patient's appearing to have an unbroken memory to earliest years, there was a specific amnesia for early childhood sexual experiences

*In order to make easier the reader's empathy with the patient's own identification, the patient will be referred to as "she" for all events before the change to "he."

†Tough, male-imitating female homosexual.

and feelings. However, there were two memories from around the age of five, the first of a neighbor boy undressing the patient and their each looking at the other's genitals. This was interrupted by the patient's mother, who spanked her. The second memory at this age was trying to urinate while standing up, but "everything backfired and I realized I wasn't supposed to go in that manner." This "realization" was reinforced by watching girls urinating sitting down.

At the age of ten, a neighbor boy attempted intercourse with her, despite his being nonplussed at his inability to find a vagina. This activity caused the patient to have her first orgasm, with ejaculation.

Puberty occurred around the age of twelve, with changes in secondary sex characteristics (without the development of breasts) and an increased sexual desire, invariably directed toward girl friends and women teachers. Masturbation began not long after, with fantasies always related to girls, especially regarding breasts. Wet dreams, which were always about women, occurred one or two times a year. During adolescence, the patient had a number of crushes on school friends and teachers and had a few relationships with girls in which there was hugging and kissing but no genital contact.

During adolescence she felt nothing unusual about the fact that she had erections and emissions, assuming that this occurred with other females. She grew tall and muscular, became very interested in athletics, and was considered a superior athlete on the girls' teams in high school, although not the most proficient of all the girls. Somewhere after the age of fourteen, her feeling that her genitals were the same as other girls' but just precocious, and that their clitoris was inside and would grow out in time, became an uncertainty. In the gymnasium she gradually became aware that the other girls looked at her and would comment and giggle about her in a hidden although at times teasing manner. She soon realized that this was related to her genitalia. Although she continued friendly with these girls, she began to withdraw, feeling that the others were looking on her as a freak. As time passed, she realized that the others not only did not but probably would not develop as she did and that the others had breasts that she did not. This gave her a desperate, hopeless, trapped feeling, which persisted more or less constantly up to the time she was first seen by us.

At the age of eighteen she was discovered during a physical examination for cystitis to have unusual genitalia for a female and so was referred to a university medical center, where the external genitalia were removed. No reconstruction in the direction of a female perineum was attempted, although the patient was offered the opportunity. Following the

operation, some growth of the stump occurred. Sexual sensation disappeared in the stump for one to two years after operation but then returned.

The first overt sexual experience as an adult occurred when she was in her twenties. Since that time, she has had many affairs, some passing and some lasting for months. Except for a single, incomplete sexual experience with a man (which did not include genital contact) her adult sexual relationships were exclusively with women. This led to the patient's social and sexual life being tied up completely with homosexuals, with all social contacts occurring either in homes or in homosexual bars and clubs. The patient never questioned her being a homosexual nor did any of those with whom she associated. Had she not considered herself and been considered to be homosexual, she would not have been fully accepted in their company. The only women with whom she ever fell in love were those who were considered in homosexual circles to be "normal"; that is, they were women who dressed and acted feminine, who had been married, and most of whom had had children, and had entered into homosexual relations only in middle life. These close and affectionate relationships were few, and preferably with women somewhat older than herself.

Pertinent Medical History

From birth to the age of 18, the patient's medical history relating to her present illness is not significant. At the time of her castration, a masculine habitus and secondary sex characteristics, rudimentary testes, and rudimentary penis with first degree hypospadias were found.

It was felt at this time that the operation was indicated because the patient had been raised as a female and for that reason would necessarily have to continue so.

In our examination at the University of California at Los Angeles, the patient was observed to be a castrated male without body or facial hair, with delicate hair on the head, and masculine, although softened, body build. Results of endocrine studies were those to be expected for a male castrated at 18.* Cystoscopic examination demonstrated a verumontanum and a very small prostate. A specimen of buccal cells and a biopsy of thigh tissue revealed male chromatin staining.

The psychiatric examination was not remarkable except for evidence of problems in psychosexual identification, in the absence of abnormal findings. The patient was an intelligent, cooperative, warm and friendly person who showed no evidence of

*17 ketosteroids and 17-Hydroxycorticoids are normal but pregnanetriole is slightly elevated. (17 ketosteroids: 17.8 milligrams; normal 6-15 milligrams. Pregnanetriole: 3.9 milligrams; normal up to 3 milligrams. 17-Hydroxycorticoids (Glenn-Nelson): 5.7 milligrams; normal 2-6 milligrams. Follicle stimulating hormone: greater than 80.)

latent or overt psychosis. Whenever the patient displayed affect, it was quite appropriate—for example, sadness when discussing a long life of being widely different from normal people, or joy with the firming up of sexual identification in the latter part of her discussions with us.

Psychological Testing

Tests employed were the Minnesota Multiphasic Inventory, Rorschach, Thematic Apperception, Sentence Completion Test, Wechsler Adult Intelligence Scale, Bender Visual-Motor Test, and a Q Sort (devised by the authors and to be reported on fully in another publication*). These revealed a person of superior intelligence and imagination, well able to use these potentials despite feelings of clumsiness, inadequacy and ineptness. Many perceptions with anxiety of mangling or destruction of the body and concern about body function were present as if this were a freshly experienced threat. There was no evidence adequate to identify the patient as homosexual.

In day to day adjustments, the patient was revealed as mildly anxious, ruminative, reserved in social interpersonal relationships but competent to deal effectively with them. Diagnostically, despite the concern over castration, the patient could be considered a normal individual with anxiety features, or as a mild anxiety neurotic.

Changes in Identification

The easiest way to measure the patient's change in identification since first being seen for this research project is that in the beginning all who knew the patient, without thinking, considered her a female. Now, no one can feel the pronouns "she" and "her" appropriate. When first seen, and for some months afterward, the patient dressed in tailored slacks, brightly colored and feminine blouses with falsies underneath, and sandals, wore jewelry and lipstick, and had plucked eyebrows and long, fine, pompadoured and carefully kept, dyed, golden hair. Bit by bit, each of these was given up. He wore shoes, men's slacks, sport shirts, which, although colorful, were typical of Southern California culture. There were no more bracelets, no makeup, no dyed hair; his hair was cut short and by a barber. When going out, he wore suits and ties for the first time in his life.

The change in identification was not confined to easily modified changes in external appearance. In addition, there was a change from delicate effeminate mannerisms, in the way he spoke, walked, blew his nose, in the phrasing of words, in timber of voice and in a host of nonverbal communications which made him indistinguishable from any gentle but vigorous man. Although there were some evi-

dences of mild passive dependency, they were not sufficient even for a diagnosis of this character structure.

For the first time in his life, he became involved in an enduring relationship with a woman (who played a very significant role in assisting him in changing his identification), whom he hopes to marry if he can legally change his sex to the biologically correct one.

DISCUSSION

Although there are many aspects of interest in this case, we will restrict ourselves to only two.

The first concerns the patient's sexual identity. Should one consider him primarily identified with a homosexual or heterosexual role previous to our study? He considered himself to be a homosexual throughout his life (until the last year or so) and was likewise considered as such by his friends, by his sexual partners and by society. He identified with the mores of the homosexual, was fearful of the social consequences of such behavior, restricted his friendships almost exclusively to homosexuals of both sexes, chose his clothes on the basis of homosexual identification and was totally imbued with the homosexual milieu in which he lived for the greater part of his adult life. Thus on the one hand he has been clearly a homosexual. On the other hand, throughout this whole period he was biologically a male. It is therefore necessary, in biological terms, to consider him to have been exclusively heterosexual. With the exception of a very few transient relations with males (and none of these adult genital male contacts) and occasional dreams of this nature, he derived sexual excitement and gratification, both in fantasy and in object relationships, exclusively with females. This is not purely a semantic problem. It goes to the heart of the continuing discussion regarding problems in sexual identification: Are these profound problems in sexual identification due to constitutional causes or to identifications derived from interpersonal relationships starting from infancy and reinforced through adult life?

The material we have available does not permit an adequate answer, although it is sufficiently rich that bias on either side of the question can find comfort. The evidence is clear that from an early age, our patient, although considered a girl by his family, had some questions about his own identification. Thus from an early age he had masculine fantasies, masculine games, and took females as sexual objects. Is this to be considered the effect of subtle biological causes or can this be considered to have resulted from disturbances in the family which, in their turn, produced intrapsychic disturbance?

*Not yet scheduled for publication.

The typical "butch" female homosexual is biologically a female, imitates men and gives a history of very early homosexual leanings. Our patient had an almost identical career and was taken for a "butch" throughout his adult life. However, the quirk is that this typical "butch" is in fact biologically a male. Thus it is possible to confuse the issue of the biological etiology of homosexuality.

Our patient to all appearances was a "butch." He was different from other "butches" only in that he was biologically a male (except for the effects of castration). Thus, despite a very discrepant constitution, his sexual identity was as confused as that of a "butch." One cannot with certainty ascribe his identity primarily to his constitutional sex, since some female homosexuals take the same identity from childhood on. Nonetheless, one wonders, without adequate evidence, if this patient may not have been propelled, almost against his wishes, by his biological sex. Whether he was compelled to his sexual identifications by unconscious forces of a primarily biological nature or by unconscious forces of a primarily psychic (disturbed identifications in the family) nature has not been determined by our methods.

The second problem is that of treatment of the intersexed patient. Our patient was highly motivated, intelligent and possessed since childhood of some questions as to whether he was as normal a female as the rest of the girls he could observe. Thus, a foundation for changing to a more masculine attitude was present by the time we began working with him. Were his identifications more fixed in the direction of being a female, it is doubtful that he could have passed over to maleness so untraumatically; it is even more questionable whether he would have wanted to. It is well known that girls suffering from hyperadrenocorticism, although both chromosomally and gonadally female, will appear in all regards completely different if one was brought up as a girl and the other as a boy.⁴

It would seem wise, when one is dealing with an intersexed patient, regardless of the etiologic background, to have a careful psychological and psy-

chiatric investigation as well as a very complete physical (including endocrinological) examination. When it is determined that the person is unequivocally committed to one sex, then the greatest caution must be used before trying to disturb this commitment. However, when there is evidence that the commitment is not clearcut (as is evidenced by our patient), then an extended workup and psychiatric treatment should be invoked, with close cooperation with other specialties to assist the patient in determining on his own to which sex he would like to belong thenceforth.²

Thus, we do not fully agree with the point of view that it is advisable to transmute all intersexed patients to their somatic sex, nor do we agree with the point of view that all intersexed patients should remain in the identification which started in childhood and persisted into adulthood. We rather believe that at times the one will be indicated, at times the other, and that the essential criterion is the strength of the patient's identification with one sex or the other. It is our belief that this can only be determined with most careful psychological and psychiatric evaluation and that no plan of treatment, in intersexed children or adults, should be embarked on until the question of sexual identification is clarified.

University of California Medical Center, Los Angeles 24 (Stoller).

REFERENCES

1. Cappon, D.: Psychosexual Identification (Psychogender) in the Somatic Pseudohermaphrodite. Presented at the Annual Meeting, American Psychiatric Association, May 1958.
2. Chapman, A. H., Saslow, G., and Watson, F.: Pseudohermaphroditism, *Psychosom. Med.* 12:212-219, 1951.
3. Freud, S.: Three Essays on the Theory of Sexuality, Imago Publishing Co., London, 1949.
4. Kiefer, J. H.: Recent advances in the management of the intersex patient, *J. Urol.*, 77:528-536, 1957.
5. Money, J., Hampson, J. G., and Hampson, J. L.: An examination of some basic concepts: The evidence of human hermaphroditism, *Bull. Johns Hopkins Hospital*, 97:4, 301-319, 1955.
6. Idem.: Imprinting and the establishment of gender role, *A.M.A. Arch. Neurol. and Psych.*, 77:333-336, 1957.